Primary aortoenteric fistula following endovascular aortic repair due to type II endoleak

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An 84-year-old female was lost to follow-up after endovascular aneurysm repair at another hospital with known type II endoleak. She later presented with presyncope and hematemesis. A referral center esophagogastroduodenoscopy showed possible duodenal diverticulum. She had recurrent symptoms and repeat computed tomography scan showed air within the aortic sac. At our center, she underwent stent graft explantation and axillofemoral reconstruction for a primary aortoenteric fistula. She was discharged and is doing well 5 months postoperatively. A high degree of suspicion for aortoenteric fistula is imperative in any patient with upper gastrointestinal hemorrhage after open or endovascular abdominal aortic aneurysm repair. (J Vasc Surg 2011;54:1164-6.)

Primary aortoenteric fistula (AEF) is a direct communication between the native aorta and the overlying bowel. It is an exceedingly rare and difficult diagnosis to make, and most are associated with abdominal aortic aneurysms. Secondary AEF complicates <2% of open abdominal aortic aneurysm (AAA) repairs.[1] Less is known about the true incidence and presentation of AEF formation following endovascular aortic repair (EVAR). Existing reported cases of AEF following EVAR have described both infectious and hemorrhagic manifestations with multiple etiologic factors.[2-15] We describe a unique presentation of a primary AEF following EVAR manifested by a subacute gastrointestinal hemorrhage.

CASE REPORT

In the spring of 2006, an 84-year-old female presented to a referring hospital with abdominal pain and an 8-cm infrarenal, fusiform AAA. She was urgently treated for a symptomatic aneurysm by endovascular means. She underwent uneventful aortic endografting with a 28-mm AneuRx graft (Medtronic, Minneapolis, Minn) with bilateral iliac extension limbs (16 mm × 11.5 mm and 16 mm × 8.5 mm).

Interval computed tomography angiogram (CTA) 2 weeks postoperatively was notable for a type II endoleak with stable aneurysm diameter. The patient then refused further imaging and ultimately was lost to follow-up, having last been seen in good condition at her 5-month postoperative visit.

Four years later, this now 88-year-old patient presented to medical attention at an outside facility with recurrent episodes of nausea, dizziness, and anemia over a 3-month period, requiring blood transfusion (hematocrit 22%) on one occasion. During this evaluation, she had a witnessed episode of hematemesis. Esophagogastroduodenoscopy (EGD) demonstrated an "unusual" posterior duodenal ulcer with no stigmata of recent bleeding, and a CT scan revealed an approximately 9.5-cm aneurysm sac. Concern was raised for AEF, and she was transferred to a tertiary care facility for further management.

Fig 1. Endoscopic photograph of duodenal erosion with recent hemorrhage.

At the referral center, the patient suffered no further recurrent episodes of gastrointestinal bleeding. She underwent a CT angiogram that demonstrated a type II endoleak, and repeat EGD was interpreted as consistent with a duodenal diverticulum (Fig 1). The patient was treated with aggressive acid reduction therapy and discharged to home on hospital day 5.

Three weeks later, we accepted this patient in transfer to our facility in hemodynamically normal condition for recurrent symptoms of dizziness, melena, and anemia (hematocrit 22%). The clinical history and abdominal CT findings of air within the aneurysm sac (Fig 2) confirmed the suspicion for AEF. After extensive discussion with the patient and her family in regard to the risk involved with aortic graft explantation, she was taken urgently to

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Fig 2. Noncontract computed tomography scan with air bubbles within aortic sac suggestive of aortoduodenal fistulization.

the operating room for repair in alignment with her expressed wishes.

In the operating room, she first underwent a right axillobifemoral bypass graft. Abdominal exploration revealed marked periaortic inflammation with an obvious site of fistula formation between the third portion of the duodenum and the anterior aortic sac (Fig 3). Proximal infrarenal aortic and distal common iliac artery control was established. Upon opening of the aneurysm, there was no evidence of purulence within the aortic sac. While there was bile staining on the wall of the aortic sac and on the mural thrombus, there was none on the stent graft itself. A single posteromedial lumbar vessel with robust pulsatile back-bleeding into the sac was identified, confirming type II endoleak. This was controlled with a suture ligature. There was no evidence of bleeding from the proximal or distal endograft seal zones or between graft components. Likewise, the fabric of the graft was intact. The aperture to the duodenal fistula was identified from within the aneurysm sac on the anterior wall. The graft was explanted without difficulty, and the proximal and distal aortic stumps were closed in two layers. After copious irrigation with antibiotic solution, the duodenum was repaired with 3-0 silk sutures in a single-layer inverting "Gambee" stitch.[16] The duodenal repair sites as well as the proximal aortic stump were buttressed with a pedicled flap of vascularized omentum.

She was treated with a 7-day course of broad-spectrum antibiotics (vancomycin, ciprofloxacin, metronidazole, fluconazole). She was discharged to inpatient rehabilitation on postoperative day 21 after a prolonged ileus. At her most recent 5-month postoperative visit, she continues to do well. She ambulates with a walker, and a sterile right groin seroma is being managed expectantly.

DISCUSSION

AEF formation following EVAR remains an extremely rare phenomenon with available literature limited only to case reports and small series.[1-15] Clinical suspicion remains the sine qua non of making this diagnosis due to its relative rarity and lack of a definitive diagnostic modality.

The current report is novel in that our patient presented with an intermittent subacute gastrointestinal hemorrhage related to aortoenteric fistulization that lasted nearly 3 months without rapid enough bleeding to manifest true hemodynamic compromise. We believe a persistent type II endoleak caused progressive aortic sac enlargement with

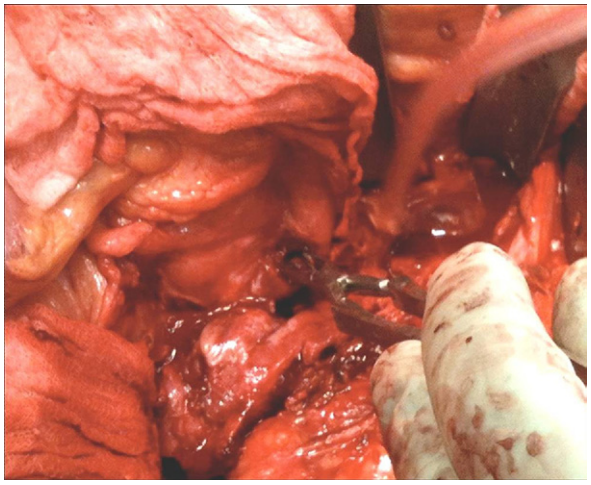

Fig 3. Intraoperative photograph. Depicted is cephalad reflection of the third portion of the duodenum with forceps within the large posterior duodenal defect.

ultimate erosion into the third portion of the duodenum. This likely manifested as a relatively low-pressure gastrointestinal (GI) bleed related to a back-bleeding lumbar artery with intermittent self-limited hemorrhage as opposed to the systemic aortic pressure seen in the more classic presentations of AEF. Additionally, the patient never manifested any systemic infectious signs or symptoms to indicate a primary graft infection. This case is best characterized as a primary AEF, as native aortic wall eroded into the duodenum from ongoing aneurysmal enlargement as opposed to a foreign body erosion/or infectious cause.

Prior case reports of AEF following endograft placement have represented more "classical" presentations of aortoenteric fistulization. Saratzis and colleagues recently reported five cases of aortoduodenal fistulization with varying endograft types. All of their patients presented with brisk hematemesis and hemodynamic compromise.[8] Earlier reports also denote the primary symptom on presentation commonly as brisk bleeding and/or shock.[7,11] In contrast, several other early series report infectious symptomatology.[3,4] A recent systematic review of reported cases of AEF following aortic endograft placement lacks specific details about each case, but did demonstrate that most patients will present with more than one complaint.[17]

The etiology of AEF formation varies widely in the reported cases as well. Reported cases account for AEF formation due to eroded translumbar and transluminal aortic coils[10,11] and type I endoleaks with sac expansion,[2,8,9,18] as well as graft kinking/erosion and primary fabric failure.[3,6,7]

This case additionally highlights the importance of patient selection for endografting as well as the concept of patient "buy-in" to maintain surveillance of these endografts postimplantation. In this case, the patient refused further radiographic imaging 6 weeks post-EVAR with a known type II endoleak and presented 4

years later with a significantly enlarged aortic sac and AEF. Persistent type II endoleaks and incomplete surveillance are both markers for poor outcomes and increased mortality following EVAR.[19,20]

EVAR is the predominant treatment modality for intact infrarenal AAA repair in the United States.[21,22] While AEF formation in the EVAR population is extremely rare, it may present in diverse ways due to myriad etiologies. As is the case after open aortic surgery, a high index of suspicion must be maintained when these patients present with gastrointestinal bleeding to avoid delay in timely intervention.

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